Received: 18 August 2020 | Accepted: 7 September 2020

DOI: 10.1002/ccr3.4163

CASE REPORT

Clinical Case Reports Open Access WILEY

# A case of obstructive jaundice without biliary stones in a low resource setting

**Elroy Patrick Weledji**[1,2] | **Dominique Ngono Ndono**[2] | **Frank Zouna**[2,3]

[1]Department of Surgery, University of Buea, Buea, Cameroon

[2]Regional Hospital, Limbe, Cameroon

[3]Department of Internal Medicine, University of Buea, Buea, Cameroon

**Correspondence**
Elroy P. Weledji, Pemset House, PO Box 126, Limbe, S.W. Region, Cameroon.
Email: elroypat@yahoo.co.uk

**Abstract**

Although not common, Mirizzi's syndrome (type 1) is a differential diagnosis in a patient with cholecystitis and obstructive jaundice, after a thorough clinical assessment. The jaundice would be resolved by cholecystectomy.

**KEYWORDS**

cholecystectomy, cholecystitis, gallbladder, jaundice, Mirizzi's syndrome, sclerotrophic

## 1 | INTRODUCTION

We present a case of obstructive jaundice without biliary stones. This was due to a scleroatrophic gallbladder causing compression (Mirizzi's syndrome) or adhesions to the extrahepatic biliary tree, pain, and recurrent cholecystitis. The jaundice resolved following cholecystectomy. Mirizzi's syndrome should be suspected in a patient with cholecystitis and obstructive jaundice.

Obstructive jaundice is a condition in which there is blockage in the pathway between the site of conjugation of bile in the liver cells and entry of bile into the duodenum through the ampulla. The extrahepatic causes are most commonly choledocholithiasis (gallstones) creating the blockage. Other causes include inflammation/ infection (primary sclerosing cholangitis, AIDS cholangiopathy etc), neoplasms (cholangiocarcinoma, pancreatic carcinoma, periampullary carcinoma, metastatic lymphadenopathy, etc), trauma, narrowing of the bile ducts (benign or malignant strictures, extrinsic compression from impacted gallbladder stone or cholecystitis (Mirizzi's syndrome), and structural abnormalities presenting at birth. The signs and symptoms of obstructive jaundice differ depending on the completeness of the blockage and the etiology. In addition, the disease course varies among individuals. A palpable enlarged healthy gallbladder (Courvoisier's law) and deepening jaundice is characteristic of a distal malignant etiology such as pancreatic carcinoma. A complete blockage typically from a common bile duct stone poses a risk of ascending cholangitis which would lead to liver failure, septicemia, and multiorgan failure. The triad of jaundice, pain, and pyrexia (Charcots' triad) is the hallmark of ascending cholangitis and requires emergency intervention.[1-3] We present a 23-year-old African man in a resource-limited setting with obstructive jaundice without biliary stones.

## 2 | CASE HISTORY/ EXAMINATION

A 23-year-old African man presented with an acute history of headache, fever, right hypochondrial pain, jaundice, and vomiting. This was associated with pruritus and the passage of dark urine and pale stools. He had no anorexia nor weight loss, nor a history of previous symptoms. His vital signs were within normal limits. He was clinically jaundiced on physical examination with tenderness in the right hypochondrium, but had no stigmata of chronic liver disease. Chest examination and cardiovascular examination were unremarkable. The preliminary differential diagnosis included acute on chronic cholecystitis, ascending cholangitis from choledocholithiasis, toxic hepatitis, a complicated malaria, or a cholangiocarcinoma. A full blood count was within normal

range (hemoglobin level of 13.4g/dl, white blood cell count of 5300/mm$^3$, platelet count of 244 000/mm$^3$) with a negative malaria parasite film. IgM antihepatitis A virus, hepatitis B surface antigen (HBsAg), antihepatitis C virus, human immunodeficiency virus (HIV) serology, and a Widal test for typhoid were negative. Direct and total serum bilirubin were markedly elevated at 26 mg/dL and 192 mg/dL, respectively, and the hepatic enzymes aspartate aminotransferase (AST) and alanine aminotransferase ( ALT) were highly elevated at 152 U/L (n 0-35) and 594 U/l(n 0-45), respectively. The biliary enzymes—alkaline phosphatase (ALP) and gamma glutamyl transpeptidase (GGT) tests to confirm extrahepatic biliary obstruction—were not available. A transabdominal ultrasound revealed a collapsed gall bladder with thick and heterogenous walls, but no gallstone or perivesicular fluid. The associated clinically obstructive nature of the jaundice gave a clinical suspicion of Mirizzi's syndrome (type 1) (Figure 1). Following an informed consent for a cholecystectomy, an upper midline abdominal incision revealed an intrahepatic chronic cholecystitic gallbladder with an inflamed Hartmann's pouch but no palpable gallstone impacting on the common hepatic (CHD)/ common bile duct (CBD). A retrograde (fundus-first) cholecystectomy allowed the disimpaction of the bladder neck from the CHD/CBD from above, and the subsequent clear dissection of Calot's triangle (bordered by the gallbladder, the common hepatic duct, and the liver). The cystic duct and cystic artery were identified with certainty, ligated, and divided close to Hartmann's pouch and the gallbladder removed. The following day the jaundice subsided. He was discharged on the 5th postoperative day with no recurrence of his symptoms at three month follow-up.

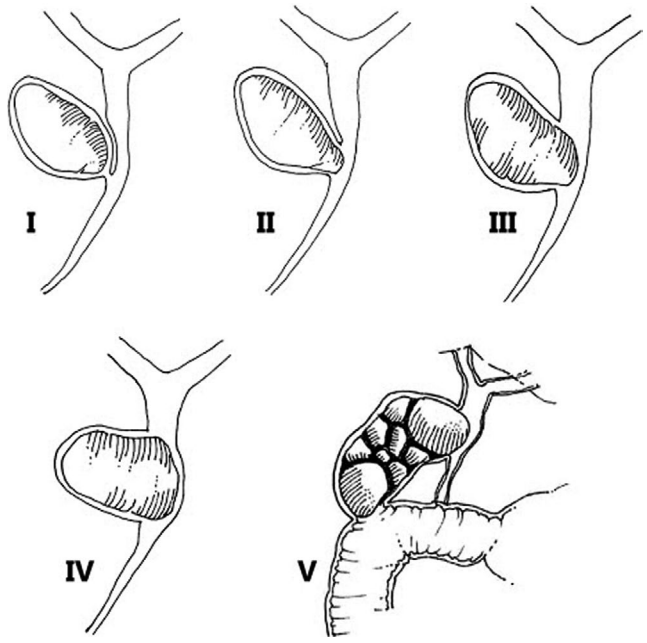


**FIGURE 1** The classification of Mirizzi's syndrome into 5 types entailing an additional type V-cholecystoenteric fistula [3] (*with permission BJS 1989;76:1139-1143)*

## 3 | DISCUSSION

This case demonstrates the rare presentation of a young African man with right hypochondrial pain associated with obstructive jaundice in a low resource setting. Clinical examination was consistent with a cholecystitis and ultrasonography revealed a collapsed gall bladder with thick and heterogenous walls, but no gallstone or perivesicular fluid. A laparotomy revealed an intrahepatic sclerotrophic gallbladder with an inflamed Hartmann's pouch extrinsically compressing the CHD/CBD but there was no palpable gallstone. The jaundice was relieved by a cholecystectomy. The etiology and differential diagnosis of the cholecystitis and obstructive jaundice is challenging. It would include (a) a sclerotrophic partially intrahepatic gallbladder which may give rise to the biliary pain and recurrent cholecystitis [4]; (b) an acute acalculous cholecystitis (acute cholecystitis without cholecystolithiasis)) manifesting as a Mirizzi's syndrome,[5-9] but for the patient having a normal white blood cell count and the presentation not meeting the full Tokyo criteria for diagnosis of acute cholecystitis.[10-12] Acute acalculous cholecystitis (AAC) is traditionally known to occur in critically ill patients and *Cyclospora cayetanensis* infection of the gallbladder in the acquired immune deficiency syndrome (AIDS) patient and has a poor prognosis.[13] However, AAC can occur in young- and middle-aged healthy individuals with a presentation not different from acute calculous cholecystitis and, the prognosis is good if diagnosed and treated early.[14] As Mirizzi's syndrome is essentially a functional hepatic syndrome elevated bilirubin and the biliary duct enzyme, alkaline phosphatase (ALP) may or may not be present, and liver function tests may not be very important [2,5] ; (c) an immunoglobulin G (IgG4)-related sclerosing cholecystitis is most commonly associated with autoimmune pancreatitis but this is not always the case[15] ; (d) the diagnosis of choledocholithiasis (CBD stone) or a biliary sludge (microlithiasis) cholecystitis [16,17] cannot be excluded because an endoscopic retrograde cholangiopancreatogram (ERCP) or magnetic resonance cholangiopancreatogram (MRCP) was not obtained and, preoperative ultrasound is notoriously not sensitive enough in detecting common bile duct stone. An intraoperative cholangiogram is important in this scenario of obstructive jaundice so as to exclude choledocholithiasis or any other bile duct pathology including a cholangiocarcinoma, but a fluoroscopic image intensifier equipment was not available; (e) although rare in this age group, the histopathology of the excised gallbladder which was unfortunately not available would have excluded gallbladder cancer and elucidated another diagnosis. From the operative aspect, it is important to note that surgery is extremely difficult in Mirizzi's syndrome as Calot's triangle is often completely obliterated

and the risk of causing injury to the CBD is high.[1,2,14] The cystic duct and artery were ligated and divided separately close to the infundibulum to avoid inadvertently ligating the CBD. This is important as the cystic duct is tented and may in addition be short or obliterated. A laparoscopic surgical approach is safe and effective with the very experienced laparoscopic surgeon.[18] In a low resource setting with minimal imaging, Mirizzi's syndrome should still be suspected in a patient with cholecystitis and obstructive jaundice. In this case, a preoperative diagnosis of Mirizzi's syndrome (type 1) led to a successful clinical outcome following a cholecystectomy.

## ACKNOWLEDGMENTS

Published with written consent of the patient.

## CONFLICT OF INTEREST

The author declares no competing interests.

## AUTHOR CONTRIBUTIONS

EPW: was the surgeon and main author; DNN: contributed to the preoperative care and literature search; FZ: contributed to the medical management and literature search.

## CONSENT FOR PUBLICATION

Written informed consent from the patient was granted to write and publish the paper.

## DATA AVAILABILITY STATEMENT

Not applicable.

## ORCID

*Elroy Patrick Weledji* https://orcid.org/0000-0002-0473-4883

## REFERENCES

1. Benjamin IS. Benign and malignant lesions of the biliary tract. In: James Garden O, ed. *Hepatobiliary and Pancreatic Surgery. A Companion of Specialist Surgical Practice*. London, UK: WB Saunders company Publishers; 2009; 201.
2. Clemente G, Tringali A, De Rose AM, et al. Mirizzi syndrome: diagnosis and management of a challenging biliary disease. *Can J Gast Hep*. 2018;2018:6962090.
3. Csendes A, Diaz JC, Burdiles P, Maluenda F, Nava O. Mirizzi syndrome and cholecystobiliary fistula: a unifying classification. *British J of Surgery*. 1989;11:1139-1143.
4. Akoglu M, Ercan M, Birol Bostanci E, Teke Z, Parlack E. Surgical outcomes of laparoscopic cholecystectomy in sclerotrophic gallbladders. *Turk J Gastroenterol*. 2010;21:156-162.
5. Shiryajev YN, Glebova AV, Korryakina TV, Kokhanenko NY. Acute acalculous cholecystitis complicated by MRCP- confirmed Mirizzi syndrome: a case report. *Int J Surg Case Rep*. 2012;3:193-195.
6. Milone M, Musella M, Maietta P, et al. Acute acalculous cholecystitis determining Mirizzi syndrome: case report and literature review. *BMC Surg*. 2014;14:90.
7. Ippolito RJ. Acute acalculous cholecystitis associated with common hepatic duct obstruction: a variant of Mirizzi's syndrome. *Conn Med*. 1993;57:451-455.
8. Mergener K, Enns R, Eubanks WS, Baillie J, Branch MS. Pseudo-Mirizzi syndrome in acute cholecystitis. *Am J Gastroenterol*. 1989;93:2605-2606.
9. Ahlawat S. Acute acalculous cholecystitis simulating Mirizzi syndrome: a very rare conditition. *South Med J*. 2009;102:188-189.
10. Kimura Y, Takada T, Strasberg SM, et al. TG13 current terminology, etiology, and epidemiology of acute cholangitis and cholecystitis. *J Hepatobiliary Pancreat Sci*. 2013;20(1):8-23.
11. Hirota M, Takada T, Kawarada Y, et al. Diagnostic criteria and severity assessment of acute cholecystitis: Tokyo Guidelines. *J Hepatobiliary Pancreat Surg*. 2007;14(1):78-82.
12. Huffman JL, Schenker S. Acute acalculous cholecystitis: a review. *Clin Gastroenterol Hepatol*. 2010;8(1):15-22.
13. Barie PS. Acute acalculous cholecystitis. In: Holzheimer RG, Mannick JA, eds. *Surgical Treatment: Evidence-Based and Problem –Oriented*. Munich: Zuckschwerdt; 2001. https://www.ncbi.nlm.n.h.govbooks/NBR6884
14. Ganpathi IS, Diddapur RK, Eugene H, Karin M. Acute acalculous cholecystitis: challenging the myths. *HPB (Oxford)*. 2007;9:131-134.
15. Kamusawa T, Tu Y, Nakayima H, et al. Sclerosing cholecystitis associated with autoimmune pancreatitis. *W J Gastroenterol*. 2006;12(23):3736-3739.
16. Shaffer EA. Gallbladder sludge: what is its clinical significance? *Curr Gastroenterol Rep*. 2001;3:166-173.
17. Ko LW, Lee SP. Biliary sludge and cholecystitis. *Best Pract Res Clin Gastroenterol*. 2003;17:383-396.
18. Kamalesh N, Prokash K, Pramil K, George T, Sylesh A, Shaji P. Laparoscopic approach is safe and effective in the management of Mirizzi'syndrome. *J Minim Access Surg*. 2015;11:216.

**How to cite this article:** Weledji EP, Ndono DN, Zouna F. A case of obstructive jaundice without biliary stones in a low resource setting. *Clin Case Rep*. 2021;9:e04163. https://doi.org/10.1002/ccr3.4163